Case Report

Management of Spinal Epidural Lipomatosis: A Report of Two Cases

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Summary

Spinal epidural lipomatosis (SEL) is a rare cause of spinal cord or dural sac compression. It is more common in males and usually affects the thoracic spine. This condition is often a consequence of excessive endogenous corticosteroid production or exogenous steroid administration for various intercurrent systemic diseases. The case histories of two patients suffering from lumbar SEL are reviewed and the management of the patients discussed, we are encouraged to report two cases. They are benign lesions, frequently presenting low back pain or neurogenic claudication, should be considered in the differential diagnosis, in patients with low back pain and radiculopathy. Conservative treatment should be attempted with obesity, steroid use and endogenous steroid disease, provided that there are no neurological deficits. Surgical treatment with SEL is the gold standard in patients with intractable pain or neurological deficit.

Key words: Spinal epidural lipomatosis, Low back pain, Neurogenic Cladication, Surgery, Conservative treatment

INTRODUCTION

Spinal epidural lipomatosis (SEL) is a rare and complex disorder that presents with epidural pathological fat tissue in the spinal vertebral canal causing spinal cord or dural sac compression. It is more common in males and usually affects the thoracic spine (12). This condition is often a consequence of excessive endogenous corticosteroid production or exogenous steroid administration for various intercurrent systemic diseases (12). In many patients, obesity has also been
implicated\(^\text{(12)}\). Only a few cases which occurred in the absence of these pre-existing conditions have been described\(^\text{(9)}\). The idiopathic cases in which no identifiable association with SEL is found constitute only 17\% of all cases\(^\text{(3)}\). The usual clinical manifestations are chronic dorsal or lumbar pain with progressive paresthesia and weakness in the lower limbs.

As such pathological entity requires differential diagnostic considerations and a concept for optimal treatment management. Clinical consideration and difficulties in managing are discussed.

**CASE PRESENTATION**

**Case 1**

The patient was an 80-year-old male and admitted with a 4-year-history of gait disturbance and neurogenic claudication. He had been CSF shunted for normal pressure hydrocephalus six years ago (Figure 1A). Gait and memory problems were resolved in one year. Four years after the operation, he admitted with back pain and lower extremity weakness with neurogenic claudication. His back pain did not resolve in spite of physical therapy and he had progressed motor deficits additionally. The patient was evaluated profoundly. Cranial neurological functions were normal. He was paraparetic (motor function 3–4/ 5), and had hypoesthesia over the L4 dermatome bilaterally. Straight leg raising tests were negative. Bilateral Patellar and Achilles tendon reflexes were hypoactive, bilateral ankle clonuses were negative. There was not any bowel or urinary incontinence.

He had a medical history of hypertension and Alzheimer's disease that was mild and under control. Laboratory evaluation, including whole blood count (WBC), electrolytes and creatinine (EUC), liver function tests, vitamin B12 and folate, urine analysis and culture, thyroid function tests, adrenocorticotropic hormone (ACTH) and cortisol, were all within the normal ranges. The patient was not obese and serum lipids within normal limits. He was not taking any steroids, and his body mass index (BMI) was 23.7 kg/m\(^2\) (weight 75 kilograms, height 178 centimeters).

Control cranial CT was clear and there were not any findings about shunt dysfunction. The patient underwent spinal radiological evaluation, because he had 2\(^{\text{nd}}\) motor neuron findings. Magnetic resonance imaging (MRI) showed a posteriorly located extradural mass lesion and erosion of the lamina of the L5 vertebra. The lesion was hyperintense on T1- and T2-weighted images. This area also showed marked contrast enhancement. He was diagnosed to have lumbar epidural lipomatosis that was compressing the dural sac and neural elements at the level of L2-L4 on MRI scan (Figure 1B).

Total laminectomy at L2 and left hemilaminectomy at L3- L4 levels were performed. Following removal of the ligamentum flavum, the dura was found compressed by unusual looking hypertrophied fat with increased vascularity which was different in appearance from regular epidural fat tissue. Epidural fat was removed totally. Histopathological diagnosis was spinal lipomatosis. The patient's weakness improved after the surgery, and he was able to walk with assistance in five days. At the end of the 2-years follow-up, his gait is almost normal and able to walk 2 hours a day without limitations. He noted, occasional dysesthesia attacks radiating from hips into the legs following prolonged sitting. He had no urinary or bowel symptoms. Neurological examination revealed full strength in the legs, a small area of hypoesthesia in the lateral right foot area. There was no evidence of recurrence in follow-up 12 months later (Figure 1C).

**Case 2**

A 73-year-old man, who had had low back pain for 15 years, presented for care because of severe pain down left leg, along
the lumbar L5 dermatome, of 1 year duration. His pain aggravated by walking or exercise. The patient had a past medical history of coronary artery disease after a coronary by-pass in 1996. There was no history of previous steroid treatments.

His BMI of 36.8 kg/m² (weight 109 kilograms, height 172 centimeters) was stable for the 5 years. The patient referred to endocrinology for endogenous steroid overproduction. All his laboratory values were within normal limits without serum lipids. Serum levels of total cholesterol and triglyceride were significantly high (serum triglyceride: 405mg/dl, LDL cholesterol: 210mg/dl).

On admission, Lasegue's sign was positive at 45° with hypoesthesia at level L5. There were no motor or reflexes changes. At the segment L4-L5, MRI showed moderate bulging intervertebral disc and the epidural lipomatosis compressing the lumbar spinal cord (Figure 2).

He was treated with analgesics and diet measures, which offered partial relief. Additionally, bed-rest was advised. During conservative treatment the patient reported significant relief of his symptoms, and 3 months later, 8 months after onset of symptoms, the radicular pain had disappeared completely, with only minimal back pain remaining. He lost weight twenty one kilograms in this period. His BMI was 29.7kg/m² (weight 88 kilograms, height 172 centimeters) after 8 months. Serum lipids were within normal limits (serum triglyceride: 154mg/dl, LDL cholesterol: 132mg/dl). Follow-up periods 1 year, the patient remained free of pain and symptoms.

Figure 1: A) A control CT after ventricle-peritoneal shunting. B) T2-weighted axial and sagittal images revealed lumbar epidural lipomatosis that was compressing the thecal sac at the level of L2-L4 (marked with arrow). C) Post-operative MRI showing gross total resection of SEL (marked with arrow). There was no evidence of recurrence the lesion.

Figure 2: The axial and sagittal views of MRI showed spinal canal compression by overgrowth adipose tissue (marked with arrow).
DISCUSSION

Idiopathic SEL was first reported by Badami and Hinck in 1982\textsuperscript{(1)}. Haddad, et al.\textsuperscript{(5)} first hypothesized that idiopathic SEL was a byproduct of obesity, with the gradual overgrowth of epidural fat resulting in compression of the spinal cord and nerves\textsuperscript{(6)}.

Spinal epidural lipomatosis is more frequent in men and is associated with exogenous steroid use in 55.3\% of cases\textsuperscript{(8)}. Obesity is the second-most common associated category with 24.5\% of cases. Cushing syndrome have been reported associated with SEL in 3.2\%\textsuperscript{(8)}. Finally, the idiopathic cases in which no identifiable association with SEL is found constitute 17\% of all cases\textsuperscript{(5)}. Obesity is a common predisposing factor\textsuperscript{(12)}. It is mostly seen in patients taking excess steroids like Cushing's disease or chronic steroid therapy\textsuperscript{(12)}. Other rare causes known to be associated with SEL include hypothyroidism, pituitary prolactinoma, Paget's disease of bone, intrathecal baclofen pump implantation\textsuperscript{(12)}. However, these have been isolated case reports and do not provide any definite etiopathogenic mechanism of excessive fat deposition\textsuperscript{(12)}.

SEL typically affects the thoracic spine and usually presents with spinal cord compression\textsuperscript{(12)}. Involvement of the lumbar leading to with cauda equina syndrome is relatively uncommon\textsuperscript{(12)}. The usual clinical manifestations consist of dorsal or lumbar pain with paresthesia and weakness in the lower limbs\textsuperscript{(8)}. Lumbar radicular symptoms and signs can be caused by a variety of diseases. Besides more frequent causes, such as disc herniation, bone stenosis, or spinal tumors, the possibility of compressing lipomatous tissue should not be overlooked. Lower-extremity weakness is the most common finding with decreased pinprick sensation and altered reflexes also frequently occur\textsuperscript{(2,10,11)}. Weakness is the only symptom in 20\% of SEL cases\textsuperscript{(8)}. To our knowledge, few cases of SEL presented with acute sphincter dysfunction and paraparesis\textsuperscript{(8)}.

MR imaging–based grading appears to be useful for making a diagnosis of idiopathic lumbar SEL. High-signal intensity on T\textsubscript{1}-weighted images and an intermediate signal on T\textsubscript{2}-weighted images are
characteristics of adipose tissue\(^7,10\). Epidural adipose tissue that has a thickness greater than 7 mm has been reported to be the diagnostic criterion for SEL\(^7,10\).

The treatment of SEL depends on the severity of the symptoms, the evolution time and the cause suspected (Figure 3). It ranges from conservative management to surgical excision consisting of laminectomy and fat debulking\(^8\). Surgical treatment is the choice treatment in patients with severe and rapidly progressive symptoms. The success rate of surgery in patients with SEL associated to exogenous steroids use is 77\%\(^8\). The medical treatment consists on weight loss, steroid taper, analgesics, bed rest and observation\(^3\). For SEL patients associated with obesity, the success rate of surgery is 66.70\% and it is 81.80\% for medical treatment by weight loss\(^3\). Laminectomy is the choice of surgical treatment and the success rate is 93.75\%\(^3\).

Decompressive surgery with fat debulking should be considered when diet therapy proves unsuccessful or when the patient suffers acute neurological deterioration. All patients in the present study and those described in the relevant literature achieved symptomatic relief after lumbar surgery\(^6\). It seems that the majority of these patients experience improvement or resolution of their neurological symptoms after surgical intervention\(^4\). Both our patients showed good prognosis after the treatments. To date, to the authors' knowledge, this is the first case reported of SEL complicating to normal pressure hydrocephalus.

In conclusion, physicians should be aware of this entity and keep it in mind in the differential diagnosis of patients with low back pain and radiculopathy, especially in the absence of disc herniation, bony stenosis, or spinal tumors. A detailed history and careful physical examination can help identify candidates that require MRI and specialty referral. SEL may be idiopathic or secondary to factors. SEL without idiopathic cases should undergo endocrinological evaluation. Conservative treatment includes bed rest, analgesics, physical therapy, and weight loss. Consideration of surgery should be based on two major factors: resistant to conservative therapy and neurological deficits. However, even if these symptoms are mild, careful management, including consideration of surgery, is needed. Surgical removal of SEL seems to be the most effective treatment option. In our opinion, conservative treatment should be attempted with obesity, steroid use and endogenous steroid disease, provided that there are no neurological deficits. Surgical treatment with SEL is the gold standard in patients with intractable pain or neurological deficit.

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